



TITLE:

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AUTHOR(S):

MIYAMOTO, YOSHIHISA; SHIINO, AKIHIKO; HANDA, JYOJI

CITATION:

MIYAMOTO, YOSHIHISA ...[et al]. Recurrent Callosal Hematoma with Atypical Moyamoya Disease. 日本外科宝函 1984, 53(5): 667-671

ISSUE DATE:

1984-09-01

URL:

<http://hdl.handle.net/2433/208799>

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症 例

Recurrent Callosal Hematoma with Atypical Moyamoya Disease

YOSHIHISA MIYAMOTO, AKIHIKO SHIINO and JYOJI HANDA

Department of Neurosurgery, Shiga University of Medical Science

(Director: Prof. Dr. JYOJI HANDA)

Received for Publication, May 31, 1984.

Non-traumatic or "spontaneous" hematomas of the corpus callosum may occasionally occur as a result of rupture of an aneurysm or an angiomatous malformation. Spontaneous intracerebral hematoma may also involve the corpus callosum in patients with brain tumors, or disseminated intravascular coagulation and other coagulopathies. However, spontaneous hematomas of the corpus callosum without such definitive etiologic factors are extremely rare. We report a case of atypical moyamoya disease who suffered recurrent hematoma of the corpus callosum with an interval of 2 years.

Case Report

This 55-year-old, previously healthy woman suddenly complained of severe headaches and lost consciousness shortly thereafter. Computed tomography (CT) scan showed a hematoma in the body of the corpus callosum, which had ruptured into the lateral ventricles (Fig. 1A). When she was transferred to our hospital 24 hours later, she was semicomatose and tetraparetic. Results of the routine laboratory tests including a coagulation profile were normal. Angiograms of the carotid and vertebral arteries on both sides showed an occlusion of the right anterior cerebral artery at its origin and a tight stenosis of the right middle cerebral artery, associated with basal moyamoya vessels and well-developed leptomeningeal anastomoses (Fig. 2). A diagnosis of atypical moyamoya disease was entertained. Her condition rapidly improved following an external ventricular drainage. An encephalomyosynangiosis was performed on the right side, and she was discharged with a residual right hemiparesis.

Two years later, however, she again noticed sudden severe headaches. On admission 2 hours later, she was confused and her neck was stiff, but no gross motor or sensory deficits were present and the results of the laboratory tests were non-contributory. CT scan again showed a hemorrhage in the genu of the corpus callosum and a hematocephalus (Fig. 1B). Angiography of the

Key words: Corpus callosum hematoma, Intracerebral hematoma, Moyamoya disease, Ventricular hemorrhage.

索引語: モヤモヤ病, 脳梁血腫, 脳内出血, 脳室内出血.

Present address: Department of Neurosurgery, Shiga University of Medical Science, Seta, Ohtsu, 520-21 Shiga-ken, Japan.



Fig. 1. Computed tomography without contrast enhancement on first (left) and second (right) admission, showing hemorrhage in the corpus callosum and ventricles.

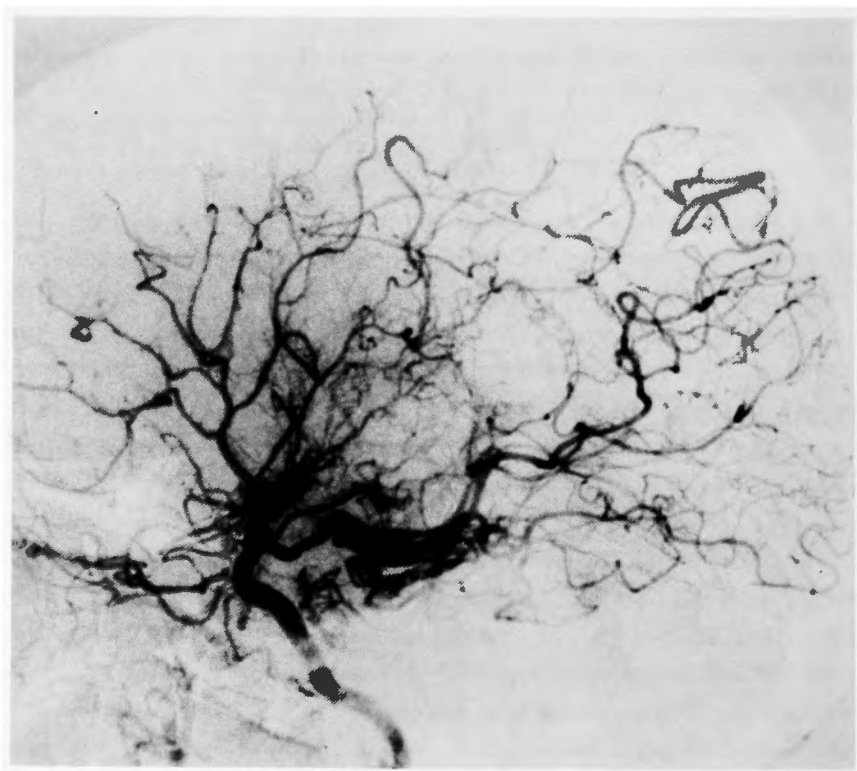


Fig. 2. Arterial phase of a lateral carotid angiogram on first admission, showing non-filling of the anterior cerebral artery and inferior group of middle cerebral branches, associated with moyamoya vessels and well developed leptomeningeal anastomoses.

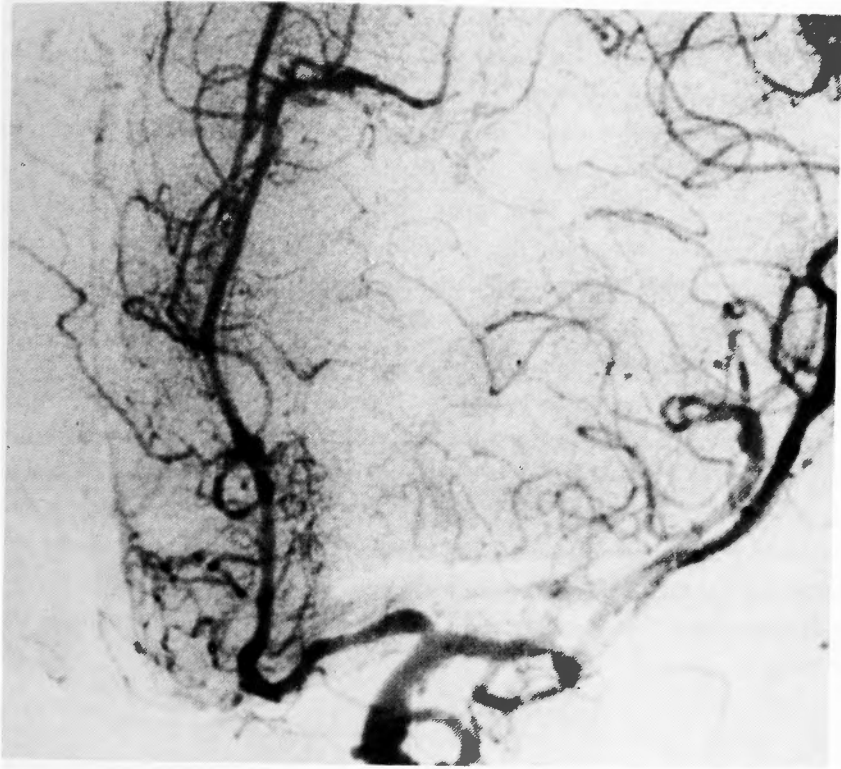


Fig. 3. Arterial phase of an oblique left carotid angiogram on second admission, showing abnormal small vessels with beaded appearance along the course of the pericallosal artery.

bilateral carotid and vertebral arteries were repeated. Several small cortical arteries were filled by right external carotid injection through the site of synangiosis. In addition, tangles of small vessels with beaded appearance were seen along the course of the pericallosal artery (Fig. 3). Her condition gradually improved and she was discharged with residual disorientation, dyscalculia, and disturbance of memory.

Discussion

The patients with moyamoya disease usually present with recurrent ischemic symptoms in children or intracranial hemorrhage in adults^{2,7}). Intracranial hemorrhage associated with moyamoya disease was once thought to be a primary subarachnoid hemorrhage. With the advent of CT scan, however, it has become known that in the majority of such cases the bleeding is primarily intraparenchymal or intraventricular.

Recently Aoki and colleagues reported 54 patients with moyamoya disease associated with intracranial hemorrhage confirmed by CT scan¹). Nine were their own and the other 45 were from the literature. They found that in all but one patient the bleeding was primarily intraparenchymal (34 patients), or intraventricular (19 patients). The single case with a primary subarachnoid hemorrhage was due to rupture of a saccular aneurysm. Our personal experiences conform

well to theirs. Three adults with moyamoya disease developed intracranial hemorrhage, which was confirmed by CT scan, operation and/or autopsy. The hemorrhage was primarily intracerebral in two and intraventricular in one⁸⁾.

True incidence of the non-traumatic hematoma of the corpus callosum remains unknown, as most previous authors reporting a large series of spontaneous intracerebral hematomas have failed to classify it as a separate group. They rarely occur primarily in the corpus callosum without well-defined causative lesions such as a ruptured cerebral aneurysm or a vascular anomaly, brain tumors, or disseminated intravascular coagulation and other coagulopathies.

It is to be noted here that Murakami and colleagues⁵⁾ recently reported a case of moyamoya disease in whom the hematoma of the corpus callosum was confirmed by CT scan. To our knowledge, however, recurrence of spontaneous hematoma in the different portions of the corpus callosum as seen in our present case has not been reported previously. Cerebral hemorrhage in patients with moyamoya disease is known to occur often from the peripheral portion of the dilated posterior or anterior choroidal artery serving as collateral channels, with or without formation of the aneurysm which is in most instances a pseudoaneurysm but rarely a true one^{3,4,6,8)}. In our present case, abnormal small vessels were present in the vicinity of the pericallosal artery. Their role in the development of the hematoma in the neighbouring corpus callosum, however, remains to be speculative.

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和文抄録

脳梁に反復して脳内血腫を生じたモヤモヤ病様疾患

滋賀医科大学脳神経外科（指導：半田譲二教授）

宮本 義久，椎野 顯彦，半田 譲二

55才，女で，2年間の間隔で2回，脳梁に特発性脳内血腫を来した症例を経験した。2回とも血腫は脳室に穿破していた。脳血管撮影で右前・中大脳動脈のつよい狭窄ないし閉塞とモヤモヤ病にいた異常な血管網の発達が見られたほか，出血源と思われる血管異常，

凝固能異常はみとめられなかった。

脳梁の出血は脳動脈瘤または動静脈奇形の破綻，あるいは腫瘍や凝固異常例で見られることがあるが，外傷例を除きまれなものとされており，更に同一例でこれを反復した例の報告はみあたらない。